Chapter 9

General discussion

Background

The aim of this thesis was to explore psychological aspects in rehabilitation. In the introduction Engel's biopsychosocial model was presented (*chapter 1*). This model adds psychological and social influences to the biomedical model and gives room for the fact that body and brain influence each other. But integration of that model in medical research seems hard since research into effects of psychological interventions in medical research is scarce. As a psychologist it was frustrating to realize that in general psychological influences on restrictions or other consequences of diseases or trauma were hardly studied, nevertheless it motivated me to find answers on the questions posed in the introduction.

Chapters

-QOL study-
Rehabilitation outpatients scored lower on all World Health Organisation Quality Of Life-abbreviated version (WHOQOL-bref) domains compared to Dutch population, thereby confirming the model of Engel [1] (*chapter 2*) but the differences on the psychological and social domain were small. The impact of chronic pain on Quality of Life (QOL) in patients was found to be higher compared to patients with musculoskeletal problems. For these two patient groups outcomes can be used as norm scores. The advantage of the WHOQOL-bref is that it provides a comprehensive overview of the adaption of the patient to their disease or trauma because all domains are assessed.
-Cognitive dysfunction study-
Rehabilitation patients, without brain damage, had higher scores on the Cognitive Failure Questionnaire compared to the Dutch population (*chapter 3*). High stress coping ability (resilience) was protecting against cognitive failure, while there was a mediating effect of anxiety and depression. The hypothesised association between cognition and surgery or pain, found in other studies, was not confirmed in our study.[2,3]
-Prosthesis satisfaction review-
The systematic review into prosthesis satisfaction (*chapter 4*) showed that satisfaction with a prosthesis was associated with many factors which could be grouped in 5 domains i.e. appearance, properties, fit, and use of the prosthesis, as well as aspects of the residual limb. Significance of the associations was related to gender, liner use, etiology and level of amputation. However, questionnaires assessing satisfaction use different operationalization's of satisfaction. In addition without providing a clear definition of satisfaction in the user guides, comparisons between study results are difficult.
-Resilience in CRPS-I study-
The mean resilience scores and mean scores on all domains of the WHOQOL-bref of patients who were amputated because of longstanding therapy resistant CRPS-I were higher than those patients with chronic pain (*chapter 5*). These results suggest that patients with resilience are capable of adapting to their new (post amputation) situation and improving their QOL.

-Association with outcome study-
Poor social support or lower score on resilience were associated with poor outcomes of an amputation in case of longstanding therapy resistant CRPS-I (*chapter 6*). These finding were somewhat disappointing because we expected associations with more psychological factors. The factors found to be associated with the outcome seem to be general factors, not specific for longstanding therapy resistant CRPS-I.
-Outcome study-
More than 75% of patients who were amputated because of longstanding therapy resistant CRPS-I reported an improvement in mobility and or a reduction in pain (*chapter 7*). Recurrence of CRPS-I occurred in the residual limb of 1 participant and in another limb of 3 participants. Despite these good result most patients still had pain and problems with their mobility. The outcomes were stable over time. These outcomes enable us to inform patients who consider an amputation for longstanding therapy resistant CRPS-I.
-Decision paper-
Because amputation in case of longstanding therapy resistant CPRS-I is controversial we described the decision making process in detail (*chapter 8*). We aimed to be transparent for patients and professionals. It forced us to reflect critically upon the decision making process and give others an opportunity to engage in a discussion about this process.

General reflection

Before the case, presented in the introduction, is discussed again in the light of our findings I will start with some more general assumptions. The result of the QOL study showed, that all domains of QOL are affected by physical problems, showing that the biomedical model is too restricted. This is in no way a new insight, it is only a confirmation of what was suggested more than 40 years ago by Engel. He noticed that the physician-patient relationship was deteriorating despite medical technical developments and innovations. The human side of illness and patient care had not developed. Additionally knowledge about the human behaviour had not been integrated: a missed chance to a more effective patient care and health maintenance.[4] Also, in the biomedical model is no room for the fact that patients want to be heard, understood and respected, and want to be involved in decision making.[5-7] These needs of patients is a development of the last decades, encouraged by health care providers.[8]
The restrictions of the biomedical model are based on Christian orthodoxy, leaving the connection with the soul, morale, mind and behavior out of this model.[4]
Nowadays we are aware of different body-brain connections in the human body e.g.: the hypothalamic-pituitary-adrenal axis, the autonomic nervous system, the immune system and the gut-brain axis,[9-13] that confirm the connection between body and brain.
This thesis is predominantly about psychological aspects of rehabilitation. It may seem that it undermines the idea of everything being interconnected by focusing on one domain. We had to select and sometimes the focus became narrow and for that

reason other aspects were ignored. In the cognitive dysfunction study for instance, we did not investigate the physical (dys)function.
In the prosthesis satisfaction review (*chapter 4*) a wide search strategy was applied to find studies investigating not only psychological domain but also the physical and the social domain. However with that broad search strategy other difficulties surfaced. A lot of different factors were associated with prosthesis satisfaction, despite the restriction of including only patients with a trans-tibial amputation. Comparison of the importance of the different factors was almost impossible due to different outcome measures (questionnaires) and study designs. In the end we just summarized all factors to get a grasp.
Another difficulty in the prosthesis satisfaction review was how factors were operationalized. Different questionnaires applied different operationalizations which were poorly documented. Some questionnaires did not have a user guide making it even more difficult to interpret results of the different questionnaires.
The last four chapters are about patients with longstanding therapy resistant CRPS-I. We encountered other difficulties in those studies. The first one was the definition of CRPS-I, which has changed a few times over the last 15 years.[14] Those changes are highly relevant when including patients in studies, determining prevalence, determining recurrence rate and when comparing outcomes of different studies.[15] But even the latest definition is under scrutiny. Some critics even state that it is not a disease at all or report overlap with other diseases and that the validity of the criteria is not sufficient and not tested thoroughly.[16,17]
A second difficulty is: how long is the diagnosis of CRPS-I valid? Most diseases have a dynamic character which is not often taken into account. For instance a person fractured a leg, becomes depressed and 10 years later developed CRPS-I which proved to be therapy resistant CRPS-I. When assessing the patient for an amputation the fracture has healed and the depression has resolved. Should we consider one of those as a factor potentially influencing the outcome of the amputation? Is the fact that the leg has fractured a sign of vulnerability? Or is the fact that the fracture has healed a sign of strength? For depression the same considerations can be made. Is depression a sign of vulnerability? Or is the fact that the patient left the depression behind a sign of strength? In general when documenting a patient's history all diagnoses are summarized but not if and how they were handled, and how the patient coped with those diagnoses. What is the value of the diagnoses 10 years after the event? I have no answers to these questions although I personally think that most of the time the way a patient handles adversity is telling me more than what kind of adversity it actually was.
The third difficulty is the goal of an amputation and interpretation of the outcome. There may be different goals a patients aims for when requesting an amputation e.g. decrease in pain, increase in mobility and or decrease infection risk. It is challenging to weigh these different goals. Is one goal more important than the other and what if a participant achieves only one of the three goals, can it be rated as an improvement or not? And if one goal is only reached for 33%, for example the pain (on a 0-10 scale) decreased from 9 to 6, how should it be rated? In my opinion even the partial realization of a goal, especially if the result improve function or QOL, is a positive

outcome. For example the decrease in pain counteracts insomnia and provides a better QOL.[18]
The fourth difficulty is a possible selection bias in our studies. Many patients with CRPS-I experience a decrease of the symptoms within 6-13 months after the onset.[19] A small group of patients progress to a longstanding therapy resistant CRPS-I. The CRPS-I patients in our studies have longstanding therapy resistant CRPS-I and differ from the other CRPS-I patients, seen by the family physician with regard to age and location of the CPRS-I.[20] The participants in the outcome study are younger and the lower limb is mostly affected. Actually our participants have more in common with samples in other studies with longstanding therapy resistant CRPS-I.[21] Younger participants and more lower limb amputations could both influence the outcome in a positive way. Younger people have more rehabilitation possibilities and the use of a lower limb prosthesis is more straightforward compared to the use of upper limb prosthesis.
Despite these difficulties the studies revealed new insights. For me it was surprising that childhood adversity and disturbing life events were not associated with a poor outcome. The relationship between adverse childhood experiences and poorer health across the life course is well established.[22-24] Additionally early adversity predisposes to chronic pain.[25] Further childhood adversity can affect the brain itself, reducing stress coping mechanism.[26,27] In CRPS-I patients stressful life events are more common, suggesting that it could be a risk factor for CRPS-I.[28] But is stress or childhood adversity influencing the outcome of an amputation in case of longstanding therapy resistant CRPS-I? We did not find such an association in the outcome study. It is possible that adversity is boosting resilience reducing the effects of adversity on outcome. Mechanism and model behind this possibility have repeatedly been investigated.[29-31] Resilience research started after the realization that childhood adversity was handled by many children in a sufficient way and the researchers wondered why.[32,33] They named the competence to handle childhood adversity: resilience. Later questionnaires were developed to measure resilience. We used the Conner Davidson resilience scale (CD-RISC) developed in 2003.[34] This questionnaire is based on different sources, for example Kobasa's work with the construct of hardiness.[35] A last remark about resilience is that although it can grow under influence of adversity the growth is not unlimited.[36]
All the difficulties and ideas concerning patients with longstanding therapy resistant CRPS-I come together in the decision making process "to amputate or not" and led to the studies of this thesis. Another way to handle as a researcher the amputation dilemma is to describe the procedure in detail and share it with others and invite them to a discussion. The process of describing the procedure forces team members to think critically about the rationale behind each step. It necessitates discussion between team members and because the team is multidisciplinary a wide view is required.
Strength and weaknesses:
The QOL study was performed in a consecutive sample of more than 500 patients and only 11% of those was excluded, making the outcomes robust. The sampling was performed in only one facility and all patients had been referred to the psychologist, reducing external validity. In the cognition study different factors were

explored in a regression model in a consecutive sample of 274 patients. However the study was limited because only one (subjective) instrument was used to measure cognition.
The outcome study included 48 participants with an amputation because of longstanding therapy resistant CRPS-I. Although the absolute sample size was small it is one of the larger samples regarding this group described in literature and part of the participants (n=17) participate in a follow up study. Limitation of that study was the lack of a control, group. However norm data found in chapter 2 helped with the interpretation of the outcomes.

Back to the case presented in the introduction of this thesis.(page 11)
Can the results of this thesis be applied in daily practice.
"Is the proposed amputation for this CRPS-I patient, a 45 year old man, the right decision?" was one of the referrals 15 years ago. At the time my mind just wondered in many different directions, not noticing that a vital part of the question was missing. The right decision for what? Are we talking about outcome? And is that the outcome the physician has in mind or the outcome the patient is aiming for? Within rehabilitation the International Classification of Functioning, Disability and Health (ICF), is used 37. The ICF model measures on 3 different levels: body structure and function e.g. edema, pain, muscle weakness; activity e.g. standing, transfers; participation e.g. work, sports. Which level are we talking about and is one of those levels more important than the other? In the outcome study different outcomes were assessed but in daily practice of the decision making process two outcomes are currently considered crucial i.e. decrease of pain and increase in mobility. Associated with these two crucial outcomes are social support and resilience. Patients who experience strong social support and who are resilient have a better chance on a good outcome after the amputation. Although the results were statistically significant in the "Association with outcome study" the model was not perfect.
Another way of looking at the referral of the rehabilitation physician is to evaluate the thinking process of the patients. Is the patient able to think clearly? Beside the cognitive side of thinking, the thinking process was discussed in the team and two crucial factors were acknowledged. Has the patient weighed the consequences of an amputation (pro and con's) thoroughly and has the patient discussed an amputation it with family or close friends? The last step in the decision making process is to compare outcome expectations of team and patient. The team discusses expectations and then compares it with the those of the patient. If the patient has a much more optimistic outcome expectation compared with that of the team, the request for an amputation is turned down.
Conclusion is that currently, if we have to decide if an amputation might be a treatment for the patient, a decision protocol is available and gives me the tools to make a decision from the psychological point of view.

Future research
The results presented in this research are a small step forward, but future research regarding the following topics is needed.
In order to improve the explained variation, presented in the association with outcome study, other psychological factors such as fear of movement, that were identified in other recent studies should be further explored.[38]
In evaluating long term outcome fixed evaluation times should be applied, for instance a measurement at 1, 2 and 5 years after amputation.
Patients who wanted an amputation but the team decided against it should be included in a follow-up study to explore outcomes in this group of patients.

General conclusions

Resilience, cognition, social support, anxiety, depression and pain are associated with outcome (QOL) of rehabilitation outpatients. Integration of psychology in (rehabilitation) medicine will not only enrich the diagnostic opportunities but also increase the therapeutic options. The dare of Engel of 40 years ago is still the same, all medical specialists increase their skills but quality of collaborations has not increased. Hence we all need to widen our view.

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